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Case Report

Stent-Assisted Coil Embolization of Ruptured Blood Blister-Like Aneurysm of the Basilar Artery: A Case Report and Literature Review

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Objective: Blood blister-like aneurysms (BBA) often develop on the anterior wall of the internal carotid artery, and few cases have been reported at other sites. We report a case of stent-assisted coil embolization in the acute phase for a ruptured BBA of the basilar artery.

Case Presentation: A 53-year-old woman underwent emergency stent-assisted coil embolization for subarachnoid hemorrhage due to a ruptured BBA in the main trunk of the basilar artery. Seven months after the operation, cerebral angiography confirmed no recurrence and a good clinical course.

Conclusion: Stent-assisted coil embolization for BBA may be one treatment option.

Keywords ▶ basilar trunk, blood blister-like aneurysm, stent-assisted coil embolization, LVIS

Introduction

Blood blister-like aneurysms (BBA) primarily develop in the non-bifurcation area of the internal carotid artery. They account for <2% of all intracranial aneurysms and <6.6% of all internal carotid artery aneurysms.[1] Other sites include the anterior communicating and basilar arteries, but only a small number of patients with BBA of the basilar artery have been reported: ruptured aneurysms, n = 16.[2–9]

The pathogenesis of BBA is thought to involve microrupture of the vascular internal elastic membrane, causing minor protrusions consisting of solely the adventitia and fibrous tissue.[4] This causes the vascular wall to become thin and fragile. When performing clipping under craniotomy or intra-aneurysmal coil embolization, the risk of intraoperative rupture is high and treatment is difficult. The standard treatment has not been established. In some patients with BBA of the internal carotid artery, parent-vessel obliteration is combined with bypass.[1] However, revascularization is difficult in those with BBA of the basilar artery; parent-vessel obliteration is impossible.

In this study, we report a patient in whom acute-phase stent-assisted coil embolization of a ruptured BBA of the basilar artery trunk led to a favorable course.

Case Presentation

The patient was a 53-year-old woman. Occipital pain developed suddenly during defecation and she was brought to our hospital by ambulance. On arrival, the blood pressure and pulse rate were 189/99 mmHg and 74 beats/min, respectively. Concerning the consciousness level, the Glasgow Coma Scale (GCS) score was E3V4M6 and the World Federation of Neurolsurgical Societies (WFNS) grade was II. There was no paralysis. She had a history of rheumatoid arthritis. She had taken methotrexate and a folic acid preparation. Head computed tomography (CT) revealed Fisher grade 3 subarachnoid hemorrhage of the right prepontine cistern (**Fig. 1A**). Magnetic resonance angiography (MRA) at the time of onset demonstrated an aneurysm-like protrusion of the basilar artery trunk (**Fig. 1C**), although no abnormal finding was found by this procedure incidentally performed 3 years previously (**Fig. 1B**). Magnetic resonance imaging

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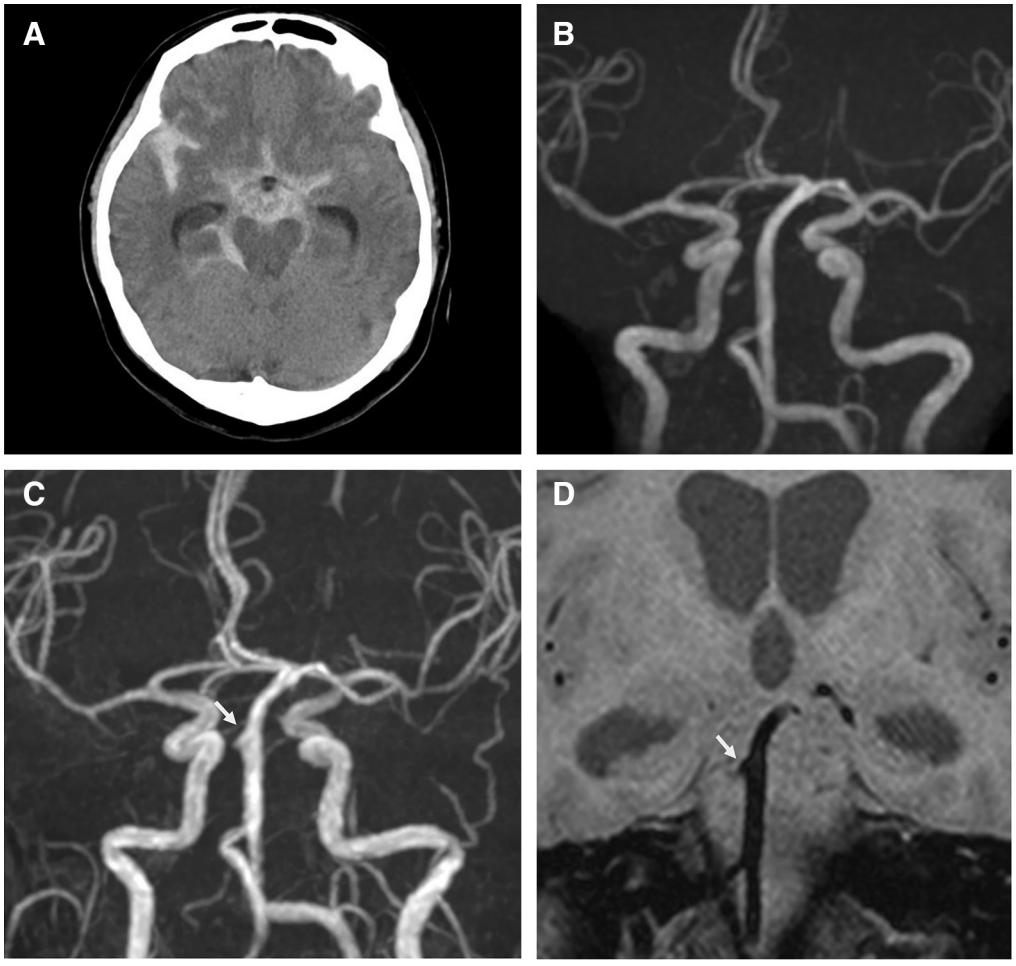

Fig. 1 (**A**) Initial head CT showed subarachnoid hemorrhage predominantly in the right prepontine cistern. (**B**) No aneurysm at the basilar trunk on MRA performed 3 years ago. (**C**) Small aneurysm (arrow) on MRA performed at onset. (**D**) Volume isotropic turbo-spin-echo acquisition (VISTA) showed a small aneurysm (arrow) and no intraluminal hematoma in the basilar artery. CT: computed tomography; MRA: magnetic resonance angiography

(MRI) T1-VISTA (Volume isotropic turbo-spin-echo acquisition, 3D-T1-TSE technique, Philips Medical Systems, Eindhoven, Netherlands) did not reveal arterial dissection (**Fig. 1D**). On emergency cerebral angiography, a rightward, wide-necked, superficial aneurysm-like protrusion measuring approximately 2.5 × 4 mm was observed in the mid portion of the basilar artery trunk (**Fig. 2A**).

Therapeutic strategy

A diagnosis of subarachnoid hemorrhage related to a ruptured BBA of the basilar artery trunk was made based on the absence of abnormality on imaging 3 years previously and morphological characteristics of the aneurysm. As a therapeutic strategy, we considered the approach to be difficult for clipping under craniotomy and the risk of intraoperative rupture to be high. On the other hand, for endovascular treatment, stent-assisted intra-aneurysmal embolization was considered to be necessary due to the wide-necked, small aneurysm. After explaining off-label use to her family and receiving informed consent, emergency stent-assisted coil embolization was scheduled. We decided to perform additional stent-assisted coil embolization or overlapping stenting after observing a change in the aneurysmal shape through a wait-and-watch period following withdrawal if intraoperative findings suggested difficulty of coil insertion.

Intraoperative findings

Under general anesthesia, 4Fr FUBUKI Dilator kits (Asahi Intecc, Aichi, Japan) were inserted into the bilateral vertebral arteries (VAs) through the bilateral femoral arteries. Heparin at 5000 units was administered. For antiplatelet drug loading, biaspirin at 200 mg and Plavix at 300 mg were administered through a nasogastric tube. Initially, for stenting, a Headway21 (Micro Vention-Terumo, Tustin, CA, USA) was inserted into the left posterior cerebral

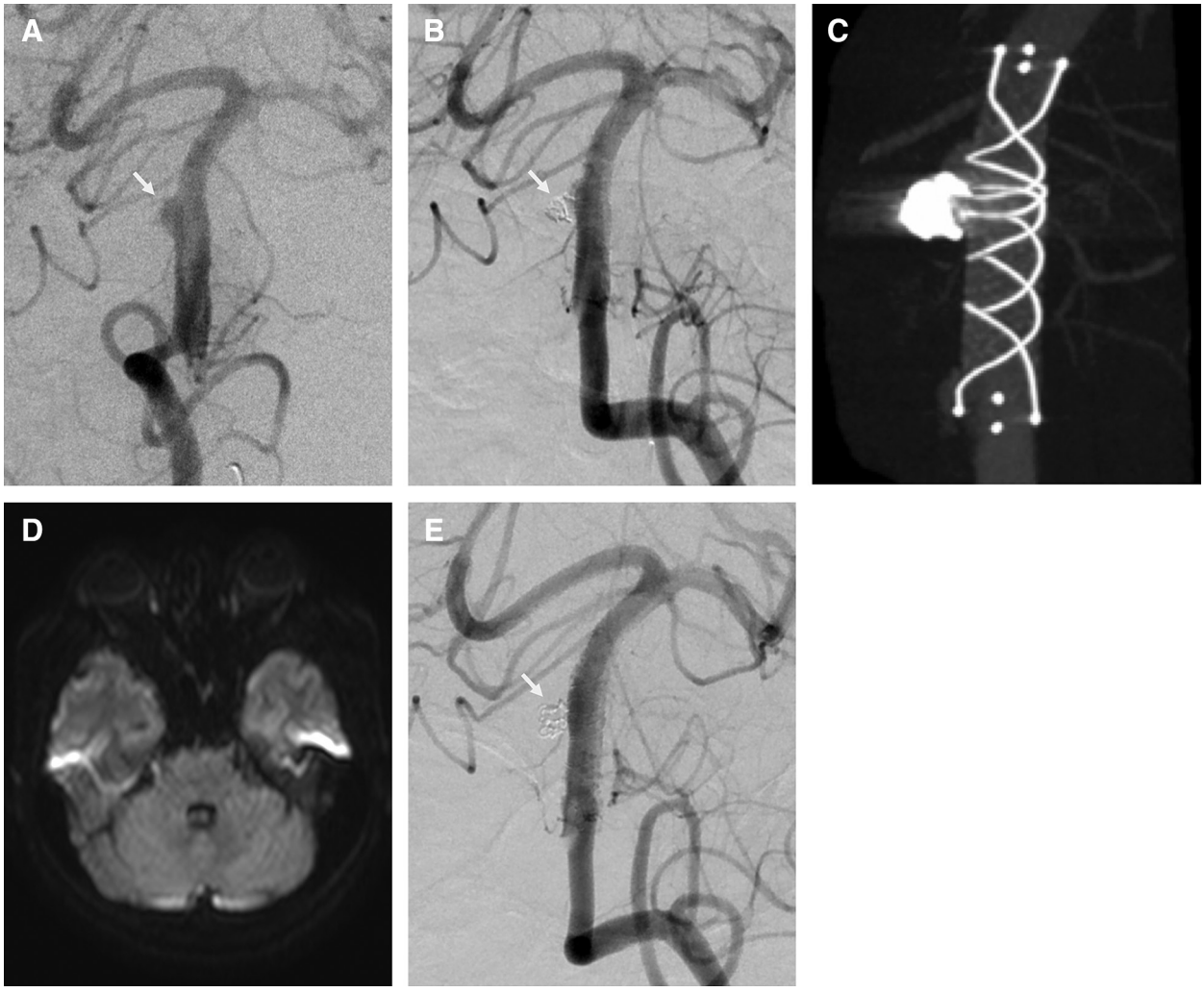

Fig. 2 (**A**) Pretreatment right VA angiography showed the small wide neck of the basilar trunk aneurysm (arrow). (**B**) Post-treatment left VA angiography showed almost complete aneurysm occlusion (arrow). (**C**) Cone beam CT showed the deployed coil and stent to be partially tight at the aneurysm. (**D**) No infarction on MRI examination on the second day after operation. (**E**) Seven-month follow-up angiography showed complete aneurysm occlusion (arrow). CT: computed tomography; MRI: magnetic resonance imaging; VA: vertebral artery

artery (PCA) coaxially with a TACTICS (Technocrat Corporation, Aichi, Japan). Subsequently, for intra-aneurysmal approaching, a double-angle steam-shaped Excelsior SL-10 preshaped 45° (Stryker Neurovascular, Kalamazoo, MI, USA) was guided using a CHIKAI10 (Asahi Intecc). An LVIS blue 4.5 × 10 (Micro Vention-Terumo, Tustin, CA, USA) was deployed using the push-and-fluff technique such that the mesh was as dense as possible at the aneurysmal site. As the 1st coil, a Target 360 ultrasoft 2.0 mm/4 cm (Stryker Neuro Vascular, Kalamazoo, MI, USA) was inserted. Subsequently, a Target 360 nano 1.5 mm/2 cm was inserted, and two Target 360 nano 1.5 mm/2 cm and 1.0 mm/2 cm were inserted. Coil deviation into the extra-aneurysmal hematoma cavity was noted and the procedure was completed. Imaging confirmed the neck remnant and the absence of an in-stent thrombus (**Fig. 2B**). Cone-beam CT involving dilution at a ratio of 5 immediately after surgery confirmed favorable stent deployment (**Fig. 2C**) and the absence of intracranial hemorrhage enlargement, and surgery was completed.

Postoperative course

MRI 2 days after coil embolization did not reveal cerebral infarction (**Fig. 2D**). There were no neurological abnormalities, and the patient commenced rehabilitation and feeding by mouth. Vasospasm was observed 10 and 11 days after surgery, and fasudil hydrochloride was administered intra-arterially. Subsequently, the course was favorable, and the patient was discharged with a modified Rankin Scale (mRS) score of 1. Cerebral angiography 7 months after surgery confirmed no recurrence and disappearance of the neck remnant related to endothelial coating (**Fig. 2E**).

Discussion

Ruptured BBA of the basilar artery trunk are rare, and the approach is difficult when opting for direct surgery. The aneurysmal shape makes treatment difficult in many cases. Pathologically, BBA are considered to be associated with dissection, differing from saccular aneurysms. Due to the pathological characteristics, the incidence of intraoperative rupture during direct surgery is high and many different surgical methods have been reported. During standard clipping of BBA under craniotomy (neck clipping), intraoperative rupture or neck avoidance may occur, making aneurysm treatment impossible in many cases. Additional hemorrhage or enlargement is observed in many patients. According to previous studies, the incidences of additional hemorrhage and enlargement after craniotomy are approximately 30% and 4%, respectively, and those after endovascular treatment are approximately 11% and 34%, respectively.[2–6,10] A previous study reported the clipping-on-crossed-wrapping method for BBA of the internal carotid artery[11] This method requires circumferential exfoliation, but it is difficult to operate on the deep area of the basilar artery; safe exfoliation at the aneurysmal periphery, which is fragile, is highly difficult. Postoperative recurrence/rebleeding has been reported. Late granulation/vascular stenosis may also develop depending on the materials used for wrapping.[12] However, this method may be more advantageous than direct clipping from the viewpoint of arterial-wall reinforcement and prevention of clip sliding.

Recent advances in neuroendovascular treatment have facilitated safe intra-aneurysmal embolization of BBA.[13] In some patients with saccular BBA, balloon-assisted intra-aneurysmal embolization is possible. Most BBA are wide-necked microaneurysms, but their shapes change over time in some cases, enabling watchful waiting until coil embolization is deemed possible.[2,3] According to previous reports, elective surgery was scheduled in most cases. However, in such cases, acute fatal rerupture may not be prevented. Furthermore, the risk of recurrence after intra-aneurysmal embolization alone is high considering the condition, and additional treatment is required in some cases.[2,3] Endovascular treatment procedures for BBA include coil embolization after waiting and watching, overlapping stenting, stent-assisted coil embolization, and stenting using a flow diverter stent.

Shah et al. conducted a meta-analysis to compare the results of treatment between craniotomy and endovascular treatment for BBA of the internal carotid artery, and reported that the aneurysmal obliteration rate was higher after craniotomy, whereas the incidence of complications after endovascular treatment was lower than after craniotomy, with a better functional prognosis; the future improvements of devices may further improve the results of treatment.[14]

The introduction of self-expanding stents for wide-necked aneurysms and parent vessel scaffold formation may prevent coil deviation into the parent vessel upon insertion of a coil into an aneurysm. Self-expanding stents exhibit flow diversion effects in addition to such scaffold effects.[15] Bulsara et al. reported treatment using a stent alone for ruptured BBA.[16] They performed stenting alone without inserting a coil into the aneurysm. However, another study found that the flow diversion effects of a self-expanding stent alone were insufficient for inducing complete obliteration.[3] Furthermore, a specific interval is required until obliteration is achieved and acute rupture cannot be prevented during this period, which is a limitation.

Walsh et al.[17] reported a treatment method in which several stents are overlapped (overlapping stenting). Flow diversion effects can be increased by overlapping several stents in a parent blood vessel. Therefore, hemodynamic aneurysmal stress may be reduced, promoting vascular endothelialization. The results of treatment with overlapping stents were favorable, suggesting the efficacy of a flow diversion effect-intended therapeutic strategy for BBA.[16,17] However, overlapping two stents may increase the risk of thrombosis. We perform prompt treatment when stent-assisted coil embolization using a single stent is possible, and perform treatment after antiplatelet drug administration when intra-aneurysmal embolization is difficult, requiring two overlapping stents, or wait for a change in the aneurysmal shape until intra-aneurysmal embolization becomes possible.

A flow diverter stent is a new endovascular treatment device for giant aneurysms. It is less porous than conventional self-expanding stents, with a higher proportion being covered by metal. Aydin et al.[4] reported 11 patients in whom treatment using a flow diverter stent for ruptured BBA was performed. In all patients, the procedure was successful and there were no acute-phase complications. Cerebral angiography at 3 and 6 months confirmed aneurysmal obliteration. In 92% of the patients, the prognosis was clinically favorable. These results suggest the efficacy of treatment using a flow diverter stent. On the other hand, another study reported that the incidence of complications was high after treatment using a flow diverter stent for posterior circulation aneurysms.[18] A reason for this may be that a flow

diverter stent is less porous than conventional self-expanding stents, to a larger degree being covered by metal; if it is inserted into the basilar artery, of which the penetrating branch count is higher than that of the internal carotid artery, penetrating-branch occlusion may cause ischemic stroke. Great attention must be paid when inserting high-metal-coverage stents, such as flow diverter and overlapping stents, in patients with BBA of the basilar artery.

Furthermore, hemodynamic stress may be promptly reduced after the insertion of a flow diverter stent, and the risk of intraoperative rupture is low in the absence of intra-aneurysmal operations, differing from stent-assisted coil embolization. However, a few weeks are required for complete thrombotic obliteration of aneurysms. During this latent period, there is a risk of aneurysmal rerupture. In patients with BBA, the risk of acute-phase rerupture is high and treatment using a flow diverter stent may be insufficient from the viewpoint of rupture prevention. Furthermore, there are problems regarding the use of antiplatelet drugs in the acute phase of ruptured aneurysms, but one previous study reported that the incidence of antiplatelet-drug-related complications after stent-assisted coil embolization of ruptured aneurysms was within a permissible range.[19] As a limitation, the use of a flow diverter stent, represented by a Pipeline, for ruptured cerebral aneurysms is currently not applicable in Japan, as described for the treatment method in the present case.

To our knowledge, 17 patients with ruptured BBA of the basilar artery, including our patient, have been reported.[2–9] In 10 patients, endovascular treatment was performed (**Table 1**). In one patient, it was combined with craniotomy. The patients' ages ranged from 44 to 68 years, with a mean of 52 years. Concerning the sex, 7 of the 10 patients were females; BBA were more frequent in females. The aneurysmal size was ≤3 mm in all 10 patients, being markedly small. They had wide-necked aneurysms. Concerning the timing of treatment, elective endovascular treatment was performed in eight patients, excluding our and another patient. The treatment methods consisted of stent-assisted coil embolization in 4 of the 10 patients, overlapping stenting in three, treatment with a flow diverter stent in two, and a combination of clipping and stenting in one patient. In a 62-year-old woman, overlapping stenting with two LVIS Jr. stents was initially conducted and an additional LVIS Jr. stent was subsequently overlapped, and coil embolization was performed. In this patient, pontine infarction occurred as a complication.[6] High-metal-coverage stent insertion into the basilar artery may increase the risk of penetrating-branch infarction.

Table 1 Cases of endovascular treatment for ruptured blood blister-like aneurysm of the basilar artery

Author/year	Age	Sex	Size (mm)	Grade (H&H)	Timing (day)	Treatment	Complications	Rebleeding	Outcome (mRS)
Meckel/2011	49	F	2.1 × 4.8	4	6	Stent-assisted coil	None	None	1
	44	F	2.0 × 3.0	3	NA	Stent-assisted coil	None	None	0
Yong/2013	45	F	NA	2	NA	Stent-assisted coil (3 stents)	None	None	0
Young/2014	53	M	2.1 × 2.0	2	Within 48 hours	Stents (Neuroform, Enterprise)	None	None	0
Michael/2014	49	F	2.5 × 1.0	1	NA	Cotton Clip + stent	None	None	1
Aydin/2015	47	F	2.0 × 2.5	1	15	Flow Diverter stent	None	None	1
	68	NA	2.5 × 4.0	2	8	Flow Diverter stent	None	None	0
Morinaga/2019	52	M	2	4	10	Stents (2 LVIS Blue)	None	None	3
	62	F	1.7	1	5, 14	Stent (2 LVIS Jr.) → stent-assisted coil (3 stents)	Infarction of right pons	None	1
Present case	53	F	2.5 × 4.0	2	1	Stent-assisted coil (1 stent)	None	None	0

H&H: Hunt and Hess; mRS: modified Rankin Scale; NA: not available

In the present case, we used a stent with strong flow diversion effects and performed intra-aneurysmal embolization. The metal coverage rate of an LVIS stent as a braided stent is higher than that of laser-cut stents; its flow diversion effects are more marked. Wang et al. reported that the flow diversion effects of a single LVIS stent were stronger than those of two overlapping stents, and that they were less marked than those of a single flow diverter stent.[20] Furthermore, the flow diversion effects of two overlapping LVIS stents were stronger than those of a single flow diverter stent.[20] When selecting stent-assisted intra-aneurysmal embolization, an LVIS stent may be advantageous for the following reasons, considering that a microcatheter may slip down (deviate from the intra-aneurysmal area) during stent deployment: this stent is highly visible, re-sheathing is possible, and re-deployment is possible through careful intraoperative operations if necessary.

As the limitations of LVIS stents, stenting-related thrombotic complications may develop and the administration of antiplatelet drugs is necessary. In addition, when coil insertion into a microaneurysm is impossible, the stent-in-stent technique must also be considered, but this may further increase the risks of thrombotic complications and penetrating-branch occlusion. Concerning stent selection for overlapping stenting, a larger number of patients should be investigated in the future. In addition, currently, LVIS stents are not applicable, and if recurrence is detected after stent insertion, intra-aneurysmal embolization may be difficult due to the high-metal-coverage rate of an LVIS stent, which is also a limitation. Internationally, similar aneurysms are often treated using flow diverter stents.

Conclusion

We reported a patient in whom stent-assisted coil embolization of a BBA of the basilar artery was performed. The use of a stent facilitates intra-aneurysmal coil embolization in some cases. Although strict follow-up is necessary, this procedure may be an effective treatment option.

Disclosure Statement

The main author completed self-reporting of conflicts of interest (COI) to the Japan Neurosurgical Society. The authors declare no COI regarding the publication of this article.

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